

Secondary Aortoduodenal Fistula

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Secondary aortoenteric fistula (SAF) is now recognized as an uncommon but exceedingly important complication of abdominal aortic reconstruction. The complication often occurs months to years after the original surgery. The main clinical manifestation of the disease is always upper gastrointestinal bleeding. Treatment of the disease is early surgical intervention. The mortality is high if no prompt operation. We present a case of secondary aortoduodenal fistula (SADF) found 20 days after aortic reconstructive surgery, with the clinical presentation of upper gastrointestinal bleeding. Even immediate exploratory laparotomy was performed, the patient died 48 hrs after the surgical management. Because of the increasing number of elective aortic aneurysm repairs in the aging population, it is likely that more patients with SAF will present to the clinical physicians in the future. So, a high index of suspicion is necessary for prompt diagnosis and treatment of this actually life-threatening event. (*Chang Gung Med* 2002;25:626-30)

Key words: secondary aortoenteric fistula, abdominal aortic reconstruction, upper gastrointestinal bleeding, aortic aneurysm, secondary aortoduodenal fistula.

Secondary aortoenteric fistulas are an uncommon but lethal complication of aortic reconstructive surgery. Because of the advancement of vascular surgery and aggressive surgical treatment, the frequency of secondary aortoenteric fistulas will likely increase. The complication often occurs months to years after the original surgery, with an incidence of 0.4%-4%.⁽¹⁾ Bastounis et al. reported that the mean interval from the initial operation to the onset of upper gastrointestinal bleeding was 32 months.⁽²⁾ The 20-year experience with secondary aortoenteric fistulas at the Johns Hopkins Medical institutions showed the average interval to be 2.8 years.⁽³⁾ The longest postoperative interval for an aortoenteric fistula reported in the literature was 27 years, in which an aortocolic fistula developed after aortofemoral bypass surgery;⁽⁴⁾ the shortest postoperative interval was 2 days, recorded in 1974, in which a parapro-

thetic-enteric fistula developed after resection of a ruptured abdominal aortic aneurysm with graft interposition.⁽⁵⁾

Dubost et al. first described homograft replacement of an abdominal aortic aneurysm in 1952.⁽⁶⁾ One year later, the first reported secondary aortoenteric fistula was reported by Brock in a case involving an aortic homograft and the duodenum.⁽⁷⁾ In 1956, Claytor et al. presented the first aortoenteric fistula caused by a prosthetic graft of the aorta.⁽⁸⁾ In 1958, Mackenzie et al. demonstrated the first successful repair of a secondary aortoenteric fistula between a synthetic graft and the intestine.⁽⁹⁾ Due to the anatomical proximity, the majority of cases involve the duodenum with the proximal suture line of an aortic prosthesis. Prompt diagnosis and surgical intervention is the only possible treatment that preserves the patient's life. Although most patients

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have so-called "herald bleeding" before fatal exsanguination, diagnosis has rarely been made before laparotomy or autopsy. In our case, the final diagnosis was made during exploratory laparotomy. Nonetheless, our case exemplifies several of the important clinical features that may lead to a diagnosis of an aortoenteric fistula preoperatively.

CASE REPORT

An 81-year-old man presented to a community hospital with a history of severe lower abdominal pain for 4 days. The severe pain was sudden in onset, and was not associated with radiation. Associated symptoms included cold sweating and dizziness. The patient had no relevant history. Because hypovolemic shock occurred at the community hospital, he was transferred to Chang Gung Medical Hospital under the impression of a ruptured abdominal aortic aneurysm (AAA). On arrival at the Chang Gung Medical Center Emergency Department, the patient was awake and alert. His blood pressure (BP) was 140/76 mmHg, with a pulse of 90 beats/minute, and a temperature of 36 °C. On physical examination, his skin was pale and he was mildly diaphoretic. Abdominal examination showed hypoactive bowel sounds, no palpable mass and no audible bruit. Laboratory investigation showed hemoglobin of 6.9 mg/dl, hematocrit of 20.5%, leukocytes of 11,600/mm³ and impaired renal function, with blood urea nitrogen of 42 mg/dl and creatinine of 2.8 mg/dl.

The patient was taken for computed tomography (CT) to evaluate whether an aneurysm existed in the abdomen. Emergency CT revealed an AAA 3 cm in diameter, with an adjacent pseudoaneurysm 6 cm in diameter. Massive hematoma was noted in the right anterior pararenal space. The patient was sent to the operating room immediately, with concomitant intravenous fluid replacement and packed red blood cell transfusion. Exploratory laparotomy showed an infrarenal AAA rupture into the right retroperitoneum with thrombi and severe atherosclerosis of the iliac artery. Resection of the ruptured AAA and interposition with a 16-mm woven graft were performed. Antibiotics therapy with cefazolin and gentamycin were chosen initially until the resected AAA culture result was reported. Cultures of the aneurysmal wall yielded *Klebsiella pneumoniae*. After the

infection doctor's consultation, ceftriaxone was administered and was suggested to use for 6 weeks instead of cefazolin and Gentamycin. On postoperative day 1, the patient's vital signs were stable, but the leukocyte count had increased to 18,400/mm³, with 19% band form. Although appropriate antibiotics were given in the following days, the band leukocyte count remained high. The patient's condition was stable with intermittent low-grade fever during the postoperative period. He felt low abdominal pain without associated symptoms or signs on postoperative day 18. On postoperative day 20, coffee ground-like material was drained from the nasogastric tube. Unfortunately, a few hours later, sudden onset of massive melena and hematemesis developed. His BP fell to 79/50 mmHg, with a pulse of 134 beats/minute. After fluid replacement, his BP returned to 112/68 mmHg. Emergency upper gastrointestinal endoscopy showed much fresh blood covering the stomach and duodenum, causing difficulty in visualizing the distal duodenum.

The patient was immediately sent to the operating room, where a huge fistula was located between the proximal anastomotic site of the aorta and the forward fourth portion of the duodenum. Profuse retroperitoneal hematoma and severe intestinal adhesion were also noted. The previous woven graft was removed. Duodenal continuity was reconstructed and an extra-anatomical vascular bypass (right axillary-bifemoral bypass) was performed.

Laboratory tests after surgery showed leukocytes of 4,900/mm³, with 25% band form. Despite appropriate antibiotics and aggressive resuscitation with blood products, crystalloid fluids and inotropic agents, a satisfactory condition and BP could not be obtained. The patient died 48 hours later after initial onset of melena and hematemesis.

DISCUSSION

A fistula between the aorta and the alimentary tract is a relatively uncommon but clinically important cause of fatal gastrointestinal hemorrhage. Secondary aortoenteric fistulas have been reported to develop from 2 days to 27 years after aneurysm repair, with a mean period from operation to diagnosis of 2.8 years.⁽²⁻⁴⁾ In our patient, a secondary aortoduodenal fistula developed 20 days later after abdominal aortic reconstruction. There have been

four cases with less than a 3-week interval, with 2 days being the shortest time to fistula development.^(9,9) This confirms the necessity of being highly suspicious of an aortoenteric fistula after aortic reconstructive surgery when a patient presents with abdominal pain, gastrointestinal hemorrhage and sepsis.

Two types of secondary aortoenteric fistulas are generally described. Type 1, termed a true aortoenteric fistula or graft-enteric fistula, with or without a pseudoaneurysm, develops between the proximal aortic suture line and the bowel. This type of fistula is the most common, and often initiates "herald bleeding" following massive gastrointestinal bleeding potentially leading to fatal exsanguinations.⁽¹⁰⁻¹²⁾ The main clinical manifestation of this type is always upper gastrointestinal bleeding (76%),⁽¹³⁾ which might be either hematemesis or melena with equal frequency. Sepsis and abdominal pain are relatively rare with this type of fistula. Type 2, or a paraprosthesis-enteric fistula, develops no communication between the bowel and the graft. It accounts for 15%-20% of secondary aortoenteric fistulas.⁽⁹⁾ In this type of fistula, bleeding occurs from the edges of the eroded bowel by mechanical pulsations of the aortic graft. Sepsis is more frequently associated with this type of fistula (57%).⁽¹³⁾ In addition to sepsis, gastrointestinal hemorrhage (30%),⁽¹³⁾ abdominal pain (20%),⁽¹³⁾ septic emboli in the lower extremities, septic arthritis, multicentric osteomyelitis and hyperostrophic osteoarthropathy have been described.^(12,14-16)

The exact pathogenesis of secondary aortoenteric fistulas is unknown, but two mechanisms have been proposed. One is the constant pulsating motion of the graft on the bowel wall, and the other is adhesion of an already infected, inflamed graft site to the wall of the gastrointestinal tract.^(5,17-19) In addition to the above mechanisms, infection or partial rupture of the suture line of the anastomosis and inadequate coverage of both the graft and proximal anastomosis with retroperitoneal tissue may contribute to SAF.⁽⁹⁾

In our case, *Klebsiella pneumoniae* was cultured from the resected AAA. The relatively high risk of either graft or suture line infection was considered. Busuttill et al., based on animal and clinical studies, proposed that a sufficient concentration of bacteria is necessary for SAF to form.⁽¹⁹⁾ Therefore, we considered that the concentration was sufficient to develop a fistula in our case because of the positive AAA cul-

ture. The most common organisms cultured are enteric pathogens and *Klebsiella* species.^(13,20) Generally, broad-spectrum antibiotic coverage following an operative procedure for SAF is necessary and ranges from 2-6 weeks according to the degree of graft infection. Patients with positive arterial wall cultures are at higher risk for subsequent disruption of the vascular anastomosis and hemorrhage and should receive 6 weeks intravenous antibiotics followed 6 months of oral administration.⁽²¹⁾ If appropriate antibiotics are administered as soon as possible to patients presenting with sepsis after mycotic aneurysmectomy with graft replacement, the infectious illness may be controlled to prevent the complication of a SAF.

Because of the nonspecific nature of the clinical history and physical findings, diagnosis of aortoenteric fistula is difficult to make preoperatively. There is no single diagnostic investigation which has a very high specificity and sensitivity, including upper gastrointestinal endoscopy, computed tomography, angiography or gallium 67 CT. Nevertheless, exploratory laparotomy is the only method that can definitely establish a diagnosis. Once the SAF is identified, the surgical procedures most commonly employed are graft excision, oversewing of the aortic stump, repair of the gut defect and placing a new graft in situ or extra-anatomic bypass.

The mortality rate during surgery and in the postoperative period is relatively high, averaging about 50%-60%.^(5,7,11) The major complications postoperatively include recurrence of the fistula, aortic stump disruption and infection of the new graft. Blowout of the aortic stump is the most common complication usually caused by residual infection. To prevent complications, an atraumatic operative technique, aseptic operative field, appropriate antibiotics and proper covering of the graft with periaortic tissue are important.

In summary, a high index of suspicion of secondary aortoenteric fistula is required in any patient who presents with sepsis, abdominal pain and gastrointestinal hemorrhage after aortic reconstructive surgery. Because of the increasing frequency of aortic reconstructive surgery, more cases of aortoenteric fistulas will be encountered in the ED. Early diagnosis and prompt surgical management are mandatory for saving the life of a patient with an aortoenteric fistula.

REFERENCES

1. Bergqvist D, Alm A, Claes G, Drott C, Forsberg O, Larsson M, Lindhagen A, Nordstrom S, Nybacka O, Ribbe E. Secondary aortoenteric fistulas: an analysis of 42 cases. *Eur J Vasc Surg* 1987;1:11-8.
2. Bastounis E, Papalambros E, Mermingas V, Maltezos C, Diamantis T, Balas P. Secondary aortoduodenal fistulae. *J Cardiovasc Surg* 1997;38:457-64.
3. O'mara CS, Williams GM, Ernst CB. Secondary aortoenteric fistula: A 20 years experience. *Am J Surg* 1981;142:203-9.
4. Shindo S, Tada Y, Sato O, Idezuki Y, Nobori M, Tanaka N. A case of an aortoenteric fistula occurring 27 years after aorto-femoral bypass surgery, treated successfully by surgical management. *Surg Today* 1993;23:993-7.
5. Elliot JP, Smith RF, Szilagyi DE. Proceedings: Aortoenteric and paraprostatic enteric fistulas. Problems of diagnosis and management. *Arch Surg* 1974;108:479-90.
6. Dubost C, Allary M, Oeconomos N. Resection of an aneurysm of the abdominal aorta: reestablishment of continuity by a preserved human arterial graft, with result after five months. *Arch Surg* 1952;64:405-8.
7. Brock RC. Aortic homografting: a report of six successful cases. *Guys Hosp Rep* 1953;102:204-28.
8. Claytor H, Birch L, Cardell Es, Zimmerman SL. Suture-line rupture of a nylon aortic bifurcation graft into the small bowel. *Arch Surg* 1956;73:947-50.
9. Mac Kenzie RJ, Buell AH, Pearson. Aneurysm of aortic homograft with rupture into the duodenum. *Arch Surg* 1958;77:965-9.
10. Connolly JE, Kwaan JH, McCart PM, Brownell D, Levine EF. Aortoenteric fistula. *Ann Surg* 1981;194:402-12.
11. Bunt TJ. Synthetic vascular graft infections. II. Graft-enteric erosions and graft-enteric fistulas. *Surgery* 1983;94:1-9.
12. O'Mara C, Imbembo AL. Paraprostatic-enteric fistula. *Surgery* 1977;81:556-66.
13. Dachs RJ, Berman J. Aortoenteric fistula. *Am Fam Physician* 1992;45:2610-6.
14. Martin A, Copeman PW. Aorto-jejunal fistula from rupture of Teflon graft, with septic emboli in the skin. *Br Med J* 1967;2:155-6.
15. Gordon SL, Nicholas GG, Garter SL, Mira AJ. Aortoenteric fistula presenting as multicentric osteomyelitis. *Clin Orthop Rel Res* 1978;131:255-8.
16. Dalinka MK, Reginato AJ, Berkowitz HD, Turner ML, Freundlich B, Steinberg M. Hypertrophic osteoarthropathy as indication of aortic graft infection and aortoenteric fistula. *Arch Surg* 1982;117:1355-9.
17. Barman AA, Kerr P. Primary and secondary aortoenteric fistula and thoracic aortic aneurysm. *NY State J Med* 1992;92:156-8.
18. Grigsby WS, Eitzen EM, Boyle DJ. Aortoenteric fistula: A catastrophe waiting to happen. *Ann Emerg Med* 1986;6:731-4.
19. Busuttil RW, Rees W, Baker JD, Wilson SE. Pathogenesis of aortoduodenal fistula: experimental and clinical correlates. *Surgery* 1979;85:1-13.
20. Umpleby HC, Turnbull AR. Arterioenteric fistulas. *Br J Hosp Med* 1988;39:488-96.